

## Treatment Outcomes in Four- to Seven-Year-Old Patients with Cleft Lip and Cleft Palate in Tawanchai Center, Srinagarind Hospital: Fistula Incidence after Cleft Palate Repair

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**Background:** Oronasal fistulae is an undesirable complication of cleft palate repair. A fistula may affect speech, eating, and socialization.

**Objective:** The purpose of the present study was to determine the prevalence of fistulae in patients who underwent primary cleft palate repair in Tawanchai Center, as well as to determine the factors that influence to fistulae.

**Materials and Methods:** A retrospective review was conducted of 96 consecutive patients between the ages of four and seven years who underwent primary cleft palate repair in Tawanchai Center at Khon Kaen University's Srinagarind Hospital. Data regarding patient demographics, age at the time of primary repair, cleft type by Veau classification, cleft width, operative technique, and surgeon's year-experiences were recorded. The incidence rates of fistulae was the primary outcomes. The associations of gender, age at the time of repair, cleft type, cleft width, and surgeon's year-experiences with the incidence of fistulae were secondary outcomes.

**Results:** There were a total of 96 consecutive patients (57 boys [59.4%] and 39 girls [40.6%]). The Mean age at primary palatoplasty was 12.9 months. The mean follow-up time after repair was 76.4 months. All patients underwent primary two-flap palatoplasty. Post-surgical fistulae were 26 (27.08%). There were 20 (20.8%) patients with symptomatic fistulae that required surgical closure. Fistulae mostly occurred at the incisive foramen (13 cases). Patients with clefts more than 1.0 cm wide and Veau cleft type IV were more likely to develop post-operative oronasal fistulae (Adjusted Odds Ratio 10.29; 95% CI = 2.10 to 50.41,  $p = 0.004$ , Adjusted odds ratio 0.10; 95% CI = 0.01 to 0.97,  $p = 0.04$  respectively).

**Conclusion:** The overall number of post-surgical fistulae were moderate rate in patients who had undergone primary cleft palate repair in Tawanchai Center. Cleft width at the time of cleft palate repair and Veau cleft type were a predictive factor for the development of post-operative fistulae.

**Keywords:** Cleft palate, Cleft palate repair, Oronasal fistula

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The management of children born with a cleft palate requires a long-term approach executed by a multidisciplinary team that includes plastic surgeons, otolaryngologists, orthodontists, and speech and language pathologist<sup>(1)</sup>. The goals of cleft palate repair are closure of the communication between the oral and nasal cavities and construction of a functional velum that allows good speech production. Surgery should result in competent velopharyngeal function while ensuring minimal disruption of maxillary growth. Post-surgical development of a fistulae is an undesirable complication of cleft palate repair. An oronasal fistulae (ONF) is defined as a

failure of healing or a breakdown in the primary surgical repair of the palate with an opening between the oral and nasal cavity<sup>(2,3)</sup>. This post-surgical complication can have an adverse effect on speech and can cause nasal regurgitation of fluids<sup>(3)</sup>.

Fistulae rates after palatoplasty have been reported to range from 12 to 45%<sup>(4,5)</sup>. Several predisposing factors may affect fistulae development, including age at the time of palatoplasty<sup>(6)</sup>, cleft type<sup>(1,7,8)</sup>, surgical technique<sup>(4,9,10)</sup> and surgeon's experiences<sup>(4,7,11,12)</sup>. Oronasal fistulae can be troublesome for both patients and surgeons.

The Srinagarind Hospital Tawanchai Cleft Center was established in 1999 for the purpose of providing multidisciplinary management of patients with cleft lip and cleft palate. The most recent update to their protocol was in 2017<sup>(13)</sup>. Tawanchai Center get appointed to be Excellence center in the name of Tawanchai Excellence Center for patients with Cleft Lip Cleft Palate and Craniofacial deformities,

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Khon Kaen University.

## Objective

The primary purpose of this research was to determine the incidence of oronasal fistulae after primary surgery. An additional purpose was to assess the association of cleft severity (including cleft type and cleft width), gender, age at time of surgery, and experience of the operating surgeon with the incidence of ONF.

## Materials and Methods

A retrospective review was conducted of consecutive patients between the ages of four and seven years who underwent primary cleft palate repair in Tawanchai Center at Khon Kaen University's Srinagarind Hospital. The following data were collected for each patient: gender, cleft type, cleft width, age at the time of primary repair, surgical technique, surgeon's experiences, presence of post-surgical ONF, location of ONF, and number of fistulae-closure operations.

The Veau classification system was used to classify the extent of the cleft, (Veau 1-cleft soft palate, Veau 2- hard/soft cleft palate, Veau 3- unilateral cleft lip/palate and Veau 4-bilateral cleft lip/palate<sup>(14)</sup>).

The Pittsburgh Fistula Classification System was used to classify the fistulae into seven types: Type I-fistulae at the uvula or bifid uvula, Type II-fistulae within the soft palate, Type III-fistulae at the junction of the soft and hard palate, Type IV-fistulae within the hard palate, Type V-fistulae at the incisive foramen (the junction of the primary and secondary palate), Type VI-lingual-alveolar fistulae, Type VII-labial-alveolar fistulae<sup>(15)</sup>.

## Outcomes and analysis

The primary outcome of this study was the presence of post-operative oronasal fistulae after primary palatoplasty, and the secondary outcomes were the associations of gender, cleft type, cleft width, surgeon's experiences, and age at the time of primary palatoplasty with the occurrence of fistulae.

Data were analyzed using STATA version 14.0 software. Odds ratios with 95 percent confidence intervals were calculated using logistic regression models. A Chi-square test and Fisher's exact test were used to compare demographic and cleft characteristics. Univariate logistic regression modeling, which included gender, age at time of primary palatoplasty, Veau cleft type, cleft width, and surgeon's experiences as variables, was conducted to identify potential factors associated with fistulae formation. Results were considered statistically significant if  $p$  was  $<0.05$ .

## Results

There were 96 patients with a cleft lip and cleft palate between the ages of four and seven years who underwent primary palate repair at Srinagarind Hospital's Tawanchai Center at Khon Kaen University (57 boys and 39 girls). The mean age at the time of primary palate repair was

12.9 months (range = 9 to 22 months; standard deviation = 2.76 months). The mean follow-up time was 76.4 months. The demographic and cleft characteristics are shown in Table 1.

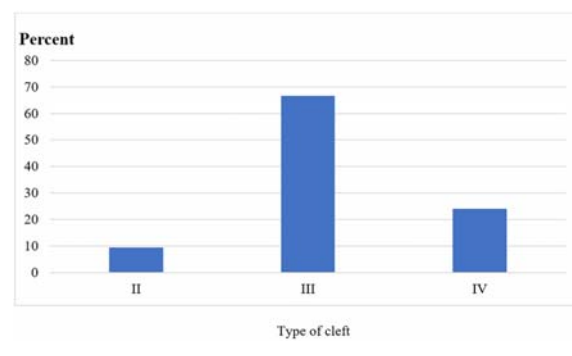
All of the 96 patients included in the study had undergone two-flap primary palatoplasty. Twenty-six (27.08%) of the patients had developed oronasal fistulae according to oral examination. Twenty of the patients had fistulae (20.83%) that were clinically significant and required surgical closure. Symptoms typically included nasal regurgitation of food and liquids and nasal escape during speech. There was also a higher incidence of fistulae in the boys 16/26 (61.53%) compared with the girls 10/26 (38.46%), but this difference was not statistically significant (Chi-square test,  $p = 0.793$ ). The majority of patients only needed one surgery to achieve successful closure of the fistula. However, one patient required a second surgery and two required a third procedure. Irrespective of the surgical technique, closing palatal fistula used a two-layer closure. Two-flap palatoplasty was used in 17 cases (85%), and a local flap was used in the remaining three (15%).

Fistulae rates according to the type of cleft was displayed in Figure 1. True fistulae formation was common in patients with unilateral cleft lip and palate (66.67%), followed by those with bilateral cleft lip and palate (23.96%). However, this was not statistically significant ( $p = 0.327$ ).

The fistulae were mostly located at the incisive foramen (type V according to the Pittsburgh Fistula

**Table 1.** Demographic and cleft characteristic of the population (n = 96)

	No.	Percentage
Sex		
Boy	57	59.38
Girl	39	40.63
Veau cleft type		
Type II	9	9.38
Type III	64	66.67
Type IV	23	23.96



**Figure 1.** Fistulae rate according to cleft type.

Classification System; 13/26 [50.0%]), followed by the junction of the hard and soft palates (type III) and in the lingual-alveolar (type VI; 4/26 [15.83%]), in the hard palate (type IV; 3/26 [11.53%]), and in the labial-alveolar (type VII; 2/26 [7.69%]). Multiple fistulae of the hard and soft palates were found in four cases.

Cleft width was the most important factor that was found to influence post-operative oronasal fistulae, that relevant to Veau cleft type. The mean cleft width was 1.22 cm with a range from 0.6 cm to 2.0 cm. According to univariate logistic regression modeling, the only factor that predicted fistulae formation was cleft width. Patients with a cleft width greater than 1.0 cm were significantly more likely to develop post-operative oronasal fistulae [OR 5.42; 95% CI = 1.48, 19.77;  $p = 0.01$ ]. Other factors (gender, Veau cleft type, age of primary palatoplasty, surgeon's experience) were not significantly associated with fistulae formation, as shown in Table 2. In multivariate logistic regression modeling cleft width and cleft severity as Veau cleft type were the predictive factors that influence to fistula formation. Patients with clefts more than 1.0 cm wide and Veau cleft type IV were more likely to develop post-operative oronasal fistulae (Adjusted odds ratio 10.29; 95% CI = 2.10 to 50.41,  $p = 0.004$ , Adjusted odds ratio 0.10; 95% CI = 0.01 to 0.97,  $p = 0.04$  respectively), as shown in Table 3.

## Discussion

The incidence of oronasal fistulae development in our study is comparable to those found in other reports published reports over the last 30 years<sup>(14)</sup>. In these reports, the occurrence of fistulae varied widely, ranging from 4.7% to 45%<sup>(1,16,17)</sup>. In this study, the overall incidence of oronasal fistulae was 26 out of 96 cases (27.08%), meaning that the rate of fistulae formation noted in this study was in moderate incidence range.

The palatal fistulae in our study most commonly developed at the incisive foramen, followed by the junction of hard and soft palates, and the hard palate, with the least amount in the labial-alveolar. This may be explained by tissue breakdown as a result of tension at the site of wound closure<sup>(18)</sup>. In several studies, cleft severity as indicated by cleft type and cleft width, surgeons' experience was a statistically significant predisposing factor for fistulae developing after palatoplasty<sup>(18,19)</sup>.

The severity of clefts, as indicated by cleft type and cleft width, were significant predictors of fistulae. Cleft types were classified according to the Veau classification. There was a higher fistula rate in patients with a cleft of the hard and soft palates than with bilateral cleft lip and palate or those with an incomplete cleft palate. As in our study, Veau cleft type IV was a statistically significant predisposing factor for fistula formation by multiple logistic regression model (Adjusted odds ratio 0.10; 95% CI 0.01 to 0.97;  $p = 0.04$ ). In a retrospective study, Lu Yong et al<sup>(18)</sup>, examined the effect of cleft type on fistulae rate when the repair was conducted using Sommerlad's repositioning of the levator veli palatine technique. Higher fistulae rate in patients with clefts of the

**Table 2.** Univariate logistic regression modeling of factors associated with fistulae formation

	Adjusted odds ratio	95% CI	p-value
Gender			
Boy	1.25	0.47 to 3.28	0.65
Girl	1		
Age of primary repair			
≤1 year	1.33	0.52 to 3.41	0.54
>1 year	1		
Veau cleft type			
Type II	1		
Type III	0.78	0.17 to 3.46	0.74
Type IV	0.55	0.10 to 3.05	0.49
Cleft width			
≤1.0 cm	1		
>1.0 cm	5.42	1.48 to 19.77	0.01
Surgeon			
≤5 year	2.13		
>5 year	1	0.84 to 5.35	0.10

**Table 3.** Multivariate logistic regression modeling of factor associated with fistula formation

	Adjusted odds ratio	95% CI	p-value
Male	2.18	0.70 to 6.79	0.17
Age of primary repair (≤1 year)	1.61	0.56 to 4.63	0.37
Veau cleft type			
Type III	0.23	0.03 to 1.67	0.14
Type IV	0.10	0.01 to 0.97	0.04
Cleft width (>1 cm)	10.29	2.10 to 50.41	0.004
Surgeon			
≤5 year	2.47	0.90 to 6.74	0.07

hard and soft palates (21%) and bilateral cleft lip and palate (10%) than in patients with a cleft of the soft palate only (3%) or unilateral cleft lip and palate (3%). This outcome was also confirmed in several other studies<sup>(1,17,19)</sup>. Cleft width also had the effect on incidence of fistulae in terms of the extent of clefting. It is postulated that as the severity of cleft increases, there is a higher occurrence of fistulae because of excessive tension on the flaps used for closure, which can lead to microischemia of the margins with necrosis breakdown. There are several other reasons for fistulae formation including hematoma, infection, coughing secondary to anesthesia effects, the child putting either fingers or objects into the mouth<sup>(20)</sup>. In our study, cleft width was a statistically significant predisposing factor for fistulae developing after palatoplasty (Adjusted odds ratio 10.29; 95% CI 2.10 to 50.41;  $p = 0.004$ ).

The optimal age to close the palate is also controversial, and the benefits of speech development have to be weighed against the possible risks of disturbing facial

growth<sup>(21)</sup>. Some authors have advocated closure of the cleft lip and soft palate before nine months of age, and of the hard palate at 12 to 14 months of age<sup>(22)</sup>. Conversely, a one-stage palatoplasty (simultaneous closure of defects in both the hard and soft palates) should not be conducted until the patient is between 12 and 18 months of age<sup>(23)</sup>. At Tawanchai Center, the lip is repaired at three to six months of age, and recommend repairing the entire palate at 10 to 18 months to avoid possible disturbances in maxillofacial growth. However, there was no significant association between fistula occurrence and age at the time of palatoplasty in this study, a result that is consistent with those of several other studies<sup>(24)</sup>. A comparison was also made between sexes with regard to the incidence of fistulae. Several studies have shown no significant difference between the genders with respect to fistula formation<sup>(17,25)</sup>. This finding was also confirmed by this study.

In addition, surgeon's experience is a critical factor in achieving consistently favorable outcomes. Salyer et al<sup>(26)</sup> postulated significantly lower fistulae rates in the second decade of surgeons' careers. The reason may be that surgical technique is the most important factor in reducing fistulae rates. Gentle handling of the cleft margin, which demands skill during the velar muscle dissection, and minimizing tension are both important in preventing tension and injury at the site of repair. However, in this study, there was no statistically significant correlation between fistulae occurrence and surgeon's experience. A reasons may be a few cases with more severity cleft repair were performed by the senior surgeon.

If an oronasal fistulae occurred in the early stages after primary palatoplasty, treatment of fistula is conservatively, as, in most cases, small fistulae they spontaneously narrow or even close. Conservative treatment is also indicated for and asymptomatic fistulae<sup>(27,28)</sup>. In this study six patients were successfully treated conservatively.

### Limitations

In the retrospective review of patient records, the identification of small fistulae can be difficult. Sometimes a palatal fistulae that appeared as a pinhole was not considered clinically relevant. Thus, some missed small fistulae might have affected to overall rate.

### Conclusion

The rate of fistulae in Tawanchai Cleft Center was 27.08%. Cleft width at the time of cleft palate repair and Veau cleft type were a predictive factor for the development of post-operative fistulae. This will serve as a foundation for ongoing studies aimed at improving the treatment outcomes for cleft patients.

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their support in data collection, foundation and publication for this study.

### What is already known on this topic?

The management of children born with a cleft lip and cleft palate requires a multidisciplinary team approach. The primary outcomes after primary palatoplasty are related to speech. One undesirable complication is oronasal fistulae.

### What this study adds?

We report the incidence of post-surgical oronasal fistulae after primary palatoplasty in Tawanchai Center was moderate incidence and that cleft width is a predictive factor in the development of post-operative fistulae.

### Potential conflicts of interest

The authors declare no conflicts of interest.

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